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AUTHOR(S):

MIZAWA, IKUO

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EXPERIMENTAL SPASMODIC TORTICOLLIS IN CAT

by

IKUO MIZAWA

From the 1st Surgical Division, Kyoto University Medical School
(Director : Prof. Dr. CHISATO ARAKI)

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INTRODUCTION

Spasmodic torticollis may be clinically defined as the abnormal involuntary contraction of the neck muscles resulting in forcing the chin in one direction with the slight tilting of the head. However, human studies have not yet shown any convincing focal pathological lesions as the cause of spasmodic torticollis, and the disagreement still continues as to whether the basic etiology is actually organic or purely functional. On the other hand, there are many evidences that spasmodic torticollis may be resulted from an organic lesion of the extrapyramidal system, and may be sometimes aggravated by psychogenic factors. Moreover, some cases of spasmodic torticollis may be truly regarded as a circumscribed form of dystonia musculorum deformans (HERZ and GLASER, 1949²¹⁾; HERZ and HOEFER, 1949²²⁾) or generalized athetosis (FOERSTER, 1929¹³⁾), or sometimes as a sequela of epidemic encephalitis (LÉVY and WIMMER).

Considering these facts, it may be considered that some cases of spasmodic torticollis may be benefited by destruction or inactivation of the selected portions of the basal ganglia.

The present study is concerned with following problems; namely, (1) whether

spasmodic torticollis is produced experimentally by making lesions in the central nervous system, (2) if spasmodic torticollis is produced, whether such an abnormal posture of the head is improved by destruction of the selected portions of the basal ganglia, and (3) destruction of which nuclei of the basal ganglia and how extensive lesions are the most effective.

PART I

SPASMODIC TORTICOLLIS-LIKE POSTURE

EXPERIMENT I

LOCATION OF A LESION ESSENTIAL TO PRODUCTION OF EXPERIMENTAL SPASMODIC TORTICOLLIS

The pathological changes in cases of spasmodic torticollis have been shown to be widespread in the central nervous system. They consist of the diffuse chronic changes of cells in the frontal lobe, the putamen, the pallidum, the thalamus, the hypothalamus, the mesencephalon and/or the vestibular nuclei. (FOERSTER, 1933¹⁴) ; GRINKER and WALKER, 1933¹⁶) ; ALPERS and DRAYER, 1937¹¹) ; HYDMAN, 1939²⁶) ; PATTERSON and LITTLE, 1943⁴³).

However, only several reports have been published regarding the production of the cephalic deviation in experimental animals, in all of which the lesion was made either in the mesencephalon, or in the rhombencephalon. (v. ECONOMO and KARPLUS, 1909¹⁰) ; RADEMAKER, 1926⁴²) ; NORTHINGTON and BARRERA, 1934³⁷) ; FERRARO and BARRERA, 1936¹¹) ; DOW, 1938⁹) ; FERRARO, PACELLA and BARRERA, 1940¹²) ; METTLER, 1943³³), 1944³⁴) ; KEMBERLING, BAIRD and SPIEGEL, 1952³¹) ; HASSLER and HESS, 1954²⁰) ; CARREA and METTLER, 1955⁶) ; ORIOLI and METTLER, 1956³⁸) ; FOLTZ, KNOPP and WARD, 1959¹⁵). Among these reports, FOLTZ, KNOPP and WARD reported in 1959 that "true" spasmodic torticollis was produced in 7 monkeys (*Macaca mulatta*) by making a lesion in the mesencephalic tegmentum which involved the portions of the medial reticular formation, the brachium conjunctivum and the medial longitudinal fasciculus.

Considering these reports, the location of a lesion essential to production of experimental torticollis seems to be in the mesencephalic tegmentum. The present study, therefore, was attempted to know which portion of the mesencephalic tegmentum is the most important for production of experimental spasmodic torticollis.

1. *Material and Method*

Experiments were carried out on 66 young adult cats, weighing 1.5-2.5 kg. All cats were anesthetized with Nembutal (40-50 mg/kg, intraperitoneal injection), and lesions were made stereotaxically with a direct current of 3 MA, 10-20 V, for 20 seconds through a monopolar electrode in the paramedian mesencephalic tegmentum according to the stereotaxic atlas of diencephalon of cat by JASPER and AIMONE-MARSAN. Electrodes consisted of fine stainless steel wires of 0.3 mm in diameter which were insulated with polyurethane except for a bare tip of 0.8 mm in length. These electrodes were inserted into the calvarium through a small trephine opening hole on the sagittal sinus in the parietal region. The size of a lesion was approximately 1.5 mm in diameter and 4.0 mm in length, so

that the decussation of the brachium conjunctivum, the medial longitudinal fasciculus and the medial reticular formation at the various levels around the red nucleus may be destroyed.

2. Results

i) Lesion at the region caudal to the red nucleus

Torticollis was produced in 13 (marked : 2, moderate : 2 and slight : 9) of 39 cats (33.3%).

ii) Lesion at the level of the most caudal pole of the red nucleus

Torticollis was produced in 12 (marked : 3, moderate : 2 and slight : 7) of 19 cats (63.2%).

iii) Lesion at the level of the magnocellular part of the red nucleus

Torticollis was produced in 7 (marked : 3, moderate : 1 and slight : 3) of 8 cats (87.5%).

Above-mentioned results indicate that the most effective lesion for production of spasmodic torticollis-like posture of cat should involve the portion of the mesencephalic tegmentum at the level of the magnocellular part of the red nucleus.

3. Stimulation of the above-mentioned portion of the mesencephalic tegmentum

Using the implanted electrodes into the above-mentioned portion of the mesencephalic tegmentum, electrical stimulation, lasting 3-5 seconds of rectangular current pulses of 1.0-3.0 msec. duration, 3-10 V, 6-12 or 60-100 cycle/sec., was applied. With the stimulation, the head (chin) was rotated toward the side opposite the stimulation in all 5 cats. This abnormal bizarre posture of the head was almost the same as that seen in spasmodic torticollis of human. When stimuli were intensified, 3 out of 5 cases showed additional neurological signs, such as anisocoria, spiral movement activity, forced circling movement, extension of the forelimb on the same side of the stimulation and, occasionally, slight flexion of the forelimb on the opposite side. However, neither tremor nor nystagmus was observed in any case. After the stimulation, an electrolytic lesion was made through the same implanted electrodes, resulting in spasmodic torticollis-like posture with deviated chin toward the side of the lesion.

EXPERIMENT II

PRODUCTION OF SPASMODIC TORTICOLLIS-LIKE POSTURE

In Experiment II, a lesion was made in 102 cats stereotaxically to destroy the decussation of the brachium conjunctivum, the medial longitudinal fasciculus and the medial reticular formation at the level of the magnocellular part of the red nucleus in the same way as in Experiment I. Coordination of the target points was determined by referring to the stereotaxic atlas of diencephalon (JASPER and AJMONE-MARSAN), as shown in Table 1.

Table 1

Coordination of the target points
(referring to the stereotaxic atlas by JASPER & AJMONE-MARSAN)

F.	- 5.0	- 5.0	- 4.0	- 4.0	- 3.0	- 3.0	- 2.0	- 2.0
L.	2.0	2.0	1.8	1.8	1.6	1.6	1.5	1.5
H.	-3.5	-2.5	-3.6	-2.6	-3.8	-2.8	-3.7	-2.7

Electrolytic lesion was made with a direct current of 3 MA, 10~20 V, for 20 seconds.

All the animals were carefully observed on neurological deficits for several weeks postoperatively. During the period of observation, photographs and short movies were often taken to document the spasmodic torticollis-like posture.

1. Results

Marked spasmodic torticollis-like posture was produced in 53 animals (51.0%), moderate one in 14 (13.7%), and slight one in 13 (12.7%), and the remaining 22 animals (21.6%) exhibited no spasmodic torticollis-like posture (Table 2).

In cases in which spasmodic torticollis-like posture was produced, the head was usually slightly rotated around the longitudinal axis and definitely around the vertebral axis toward the side of the lesion, and slightly overextended backwards. Such a posture of the head was accompanied by clonic and/or tonic contraction of the neck muscles except in a state of sleep (Fig. 1 a).

These conditions were seen immediately after the operation, and accentuated extremely on emotional states (Fig. 1 b). In several cases, such a cephalic rotation was so

Table 2

Production of spasmodic torticollis-like posture

	No. of cases	%
Marked	53	51.0
Moderate	14	13.7
Slight	13	12.7
No	22	21.6
Total	102 cases	100%

Table 3

Additional symptoms of 80 spasmodic torticollis-like posture

	No. of cases	%
Disturbances of equilibrium	48	68
Palsy of the 3rd cranial nerve	37	46
Motor disturbances of extremities	36	45
Spiral movement	8	10
Curvature of the body	7	9
Forced circling movement	2	2.5



Fig 1 a



Fig 2 b

Fig. 1 Cat No. 82 Four days after destruction of the left mesencephalic tegmentum. Fig 1a shows the torticollis at rest, and Fig. 1b on emotional excitations.

marked that the animals sometimes did forced spiral movements and could not eat or drink by themselves. In almost all cats with or without spasmodic torticollis-like posture, other additional neurological deficits were accompanied, such as disturbances of equilibrium, palsy of the ipsilateral 3rd cranial nerve, motor disturbances, forced circling movement and hypotonia of the extremities on the side opposite the lesion (Table 3). However, the tonic contraction of the neck muscles and other additional neurological symptoms disappeared gradually within several days postoperatively. But, spasmodic contraction of the neck muscles persisted for several weeks postoperatively until the time of sacrifice.

2. Histological examination

The cats were sacrificed several weeks postoperatively. The brains were perfused *in situ* with 200~500ml of 10% formalin solution, and removed carefully. Blocks of the all brains for microscopic study were embedded in celloidin, sectioned serially at the thickness of 20 microns and stained with the modified KLÜVER and BARRERA'S method.

i) Marked spasmodic torticollis-like posture

Histological studies were made in 14 cats in which marked spasmodic torticollis-like posture was seen. The lesion common to all 14 cats was found to involve the magnocellular part of the red nucleus, the decussation of the brachium conjunctivum, the medial longitudinal fasciculus as well as the medial reticular formation, especially the reticular formation surrounding the dorso-medial part of the red nucleus at the level of the magnocellular part of the red nucleus (Table 4; Fig. 2, 3).

ii) No spasmodic torticollis-like posture

Seven cats in which no spasmodic torticollis-like posture was observed, were examined as controls. The magnocellular part of the red nucleus and the medial reticular formation had been destroyed in all 7 animals, the medial longitudinal fasciculus in 4, the decussation of the brachium conjunctivum in 3, and the central tegmental bundle in 5 (Table 5). Of interest is that, in all 4 cases in which the medial longitudinal fasciculus had been destroyed, there was no involvement of the decussation of the brachium con-

Table 4
Extent of the lesions of the marked spasmodic torticollis-like posture

	10	20	45	50	64	78	81	97	98	101	106	107	109	117
Nucleus ruber (pars magnocellularis)	A	A	B	A	A	A	A	A	A	A	A	A	A	A
Fasciculus longitudinalis medialis	A	A	A	A	B	C	A	A	A	B	A	A	A	A
Decussatio brachii conjunctivi	A	A	A	A	A	A	A	A	A	A	A	A	A	A
Medial reticular formation	A	B	A	A	B	A	B	A	A	A	A	A	A	A
Tractus tegmentalis centralis		B	A	A	C	B	B	C	B	C	C	C	B	A
Griseum centrale	C		A	C	C	C	C	C	C	C	B	C	B	A
Nucleus nervi oculo-motorii			A	C	C	C		C	B	C	C	C	B	A
Tractus habenulo-peduncularis			A	C	C	C		B		A	B		A	
Commissura posterior						C		B	B	C				C
Nucleus commissurae posterioris						C			C	C		C	C	B
Lemniscus medialis					B	C		C			C	C		
Substantia nigra								C		C	C			C

Extent of the destruction A : almost complete B : about half C : partial

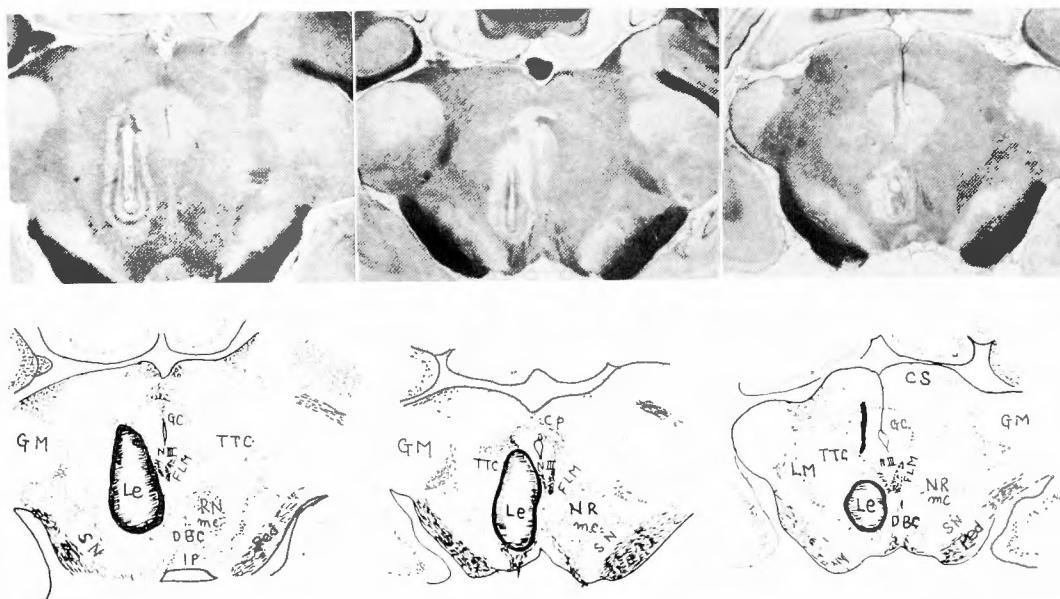


Fig. 2 Photomicrographs of cross section of the mesencephalon in cat No. 97

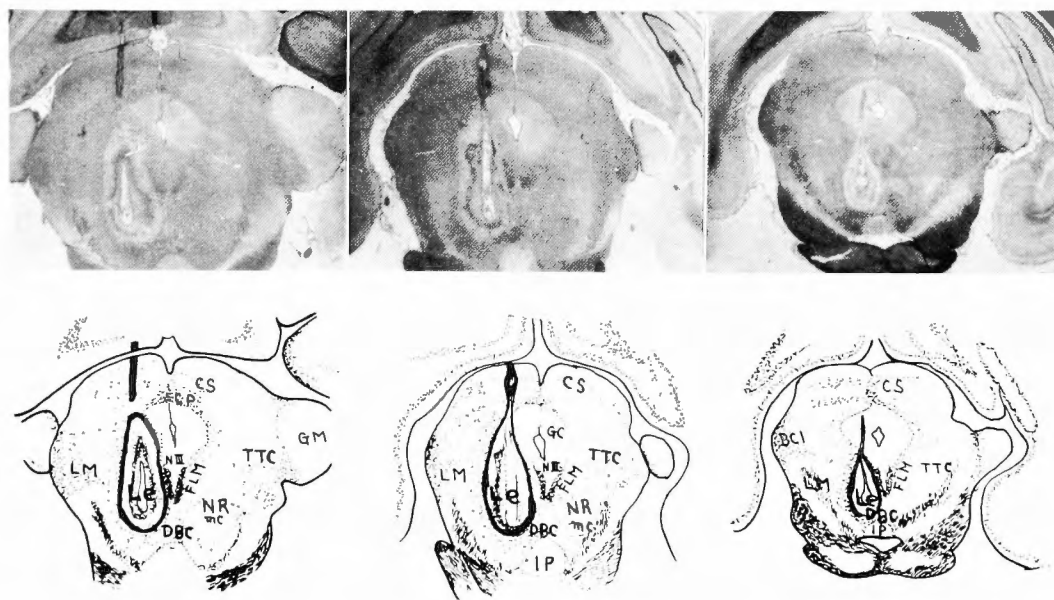


Fig. 3 Photomicrographs of cross section of the mesencephalon in cat No. 109

Table 5

Extent of the lesions of no spasmodic torticollis-like posture

	63	65	66	87	103	116	119
Nucleus ruber (pars magnocellularis)	A	A	A	A	A	A	A
Fasciculus longitudinalis medialis	C		A	A			B
Decussatio brachii conjunctivi		B			A	A	
Medial reticular formation	B	B	A	B	A	B	B
Tractus tegmentalis centralis	B	C	B		B		A
Griseum centrale	C			B		B	
Nucleus nervi oculomotorii							
Tractus habenulo-peduncularis		B	C				
Commissura posterior				C			B
Nucleus commissurae posterioris							B
Lemniscus medialis	C	A			C	B	B
Substantia nigra		B					

Extent of the destruction

A : almost complete

B : about half

C : partial

junctivum, and also in all 3 cases in which the decussation of the brachium conjunctivum had been destroyed, there was no involvement of the medial longitudinal fasciculus. Furthermore, the location of destruction either of the medial longitudinal fasciculus or of the decussation of the brachium conjunctivum in this group, was situated in the level caudal to the red nucleus.

These findings suggest that both the medial longitudinal fasciculus and the decussation of the brachium conjunctivum should be destroyed at the level of the magnocellular part of the red nucleus for production of spasmodic torticollis-like posture. Moreover, the cases, in which the medial reticular formation had been destroyed more extensively, showed usually a more marked torticollis.

COMMENT TO EXPERIMENTS I & II

A number of experimental studies have been made regarding production of abnormal posture of the head. However, the posture of the head produced experimentally in most animals was not characterized by clonic and/or spasmodic movements, but by tonically sustained deviation of the head. Among several reports (METTLER, 1944³⁴) ; ORIOLI and METTLER, 1956³⁸) ; FOLTZ, KNOPP and WARD, 1959¹⁵) ; etc.), in which clonic and/or spasmodic movements of the head were produced in experimental animals, only a paper by Foltz, KNOPP and WARD (1959)¹⁵) was concerned with "true spasmodic torticollis" in monkey. They made a lesion in the portions of the medial longitudinal fasciculus, the decussation of the brachium conjunctivum and the medial reticular formation through needle electrodes by the aid of "HORSLEY-CLARKE" apparatus. The nature of the torticollis of monkey in their report was characterized by spasmodic contraction of the neck muscles, consisting of complex clonic and tonic activity resulting in rotation and tilting of the head. Moreover, such a contraction of the neck muscles was easily accentuated and exaggerated when the animal was on emotionally excited states.

In the present study, spasmodic torticollis-like posture was produced in cat by making

a lesion in almost the same region as that FOLTZ and coworkers (1959)¹⁵⁾ reported in monkey. The posture of the head in these cats was quite similar to "dystonic" pattern of clinical cases with spasmodic torticollis.

Considering these results, the location essential to production of experimental spasmodic torticollis seems to be a region involving the medial longitudinal fasciculus, the decussation of the brachium conjunctivum and the medial reticular formation at the level of the magnocellular part of the red nucleus. Therefore, it seems to be necessary to survey the function of such nuclei and pathways from the anatomical and physiological viewpoints.

1) Medial longitudinal fasciculus---ORIOLI and METTLER (1958)⁴¹⁾ reported that only one case had the marked deviation of the head accompanied with rotated chin toward the right by destroying the right medial fasciculus alone. METTLER (1959)³⁵⁾ reported that deviation of the head was observed in 4 out of 8 monkeys in which there was the unilateral or asymmetrical lesions of the medial longitudinal fasciculus. In 3 out of 4 these animals with the cephalic deviation, the abnormal posture of the head was very marked, the chin being deviated toward the side of the lesion or to the side of a larger lesion if bilateral lesions were present. These reports seem to indicate that unbalanced activities of bilateral medial longitudinal fasciculi may play some role in production of the cephalic deviation.

2) Brachium conjunctivum---The physiological function of the brachium conjunctivum have been studied by KELLER and HARE (1934)³⁶⁾, CARREA and METTLER (1955)⁶⁾, CARPENTER and STEVENS (1957)⁵⁾, ORIOLI and METTLER (1958)⁴¹⁾ and others. KELLER and HARE (1934)³⁶⁾ pointed out that the brachium conjunctivum plays an important role for the maintenance of righting reflexes, but they did not mention the relationship between the function of the brachium conjunctivum and the deviation of the head. CARPENTER and STEVENS (1957)⁵⁾ reported that unilateral total section of the brachium conjunctivum of monkey frequently produced head tilting to the opposite side of the lesions and also caused falling and circling movement to the side of the lesion. On the other hand, CARREA and METTLER (1955)⁶⁾, and ORIOLI and METTLER (1958)⁴¹⁾ reported that unilateral lesion of the fibers of the brachium conjunctivum alone did not cause deviation of the head, but they pointed out that deviation of the head was produced in almost two thirds of experimental monkeys in which the fibers of both the brachium conjunctivum and the medial longitudinal fasciculus had been interrupted together.

These reports seem to indicate that combined destruction of the brachium conjunctivum and the medial longitudinal fasciculus may produce deviation of the head with deviated chin toward the side of the lesion. In other words, it may be postulated that damage to the fibers of the brachium conjunctivum seems to have a provocative effect rather than an essential effect for production of the cephalic deviation (METTLER, 1959³⁵⁾).

Regarding the composition of the brachium conjunctivum, many anatomical studies show that the brachium conjunctivum consists of the main fibers originating from the dentate as well as interposed nuclei and the few fibers from the fastigial nucleus of the cerebellum, although the existence of the latter few ascending fibers has been doubted in cat (CARPENTER, BRITTIN and PINES, 1958⁴⁾ ; McMASTER and RUSSEL, 1958³²⁾). Since the

deep cerebellar nuclei participate in controlling the postural reflexes and the tone of the skeletal muscles of extremities, trunk and head, it may be considered that the unilateral lesion of their projecting fibers and nuclei may produce the disturbances of equilibrium of posture, resulting in ataxia involving muscles of the extremities.

3) Reticular formation---Since the reticular formation has to do with the regulation of the impulses from the spinal centers to the upper centers, and *vice versa*, there is no doubt that the reticular formation controls over the maintenance of the tone of muscles. SPIEGEL (1925⁴⁹), 1927⁴⁸) suggested that since one of centers on static innervation of the skeletal muscles of extremities would probably exist in the reticular formation, the reticular formation might have, to some extent, to do with the production of spasmodic torticollis-like posture. Furthermore, KEMBERLING, BAIRD and SPIEGEL (1952)⁵¹) pointed out that the reticular formation may not be less important than the vestibular nuclei in controlling over the posture of the head. WARD (1947)⁵¹) also expressed his opinion that the reticular formation in the mesencephalic tegmentum may play a great role for maintaining decerebrate rigidity in monkey.

In the present study, it was confirmed that the more marked the torticollis-like posture was, the more extensive lesion was found in the reticular formation surrounding the dorsomedial part of the red nucleus.

4) Other structures---In addition to above-mentioned structures, there are some other structures which seem to be concerned with production of the abnormal posture of the head.

(a) Red nucleus : Many authors reported that a unilateral lesion restricted to the parvocellular and/or magnocellular part of the red nucleus did not always produce the deviation of the head (MUSSEN, 1927³⁶) ; INGRAM, RANSON and BARRIS, 1934²⁸) ; CARPENTER, 1956³) ; HINMAN and CARPENTER, 1959²³) ; and others). FOLTZ, KNOPP and WARD (1959)¹⁵) also reported that the red nucleus may not play the important role for production of spasmodic torticollis in monkey. However, in the present study, there was no case in which the lesion was restricted only to the red nucleus. Therefore, no conclusion can be made on the role of the red nucleus for production of spasmodic torticollis-like posture. But it was confirmed that if the red nucleus was destroyed at the level of the magnocellular part of the red nucleus, spasmodic torticollis-like posture would be more easily produced.

(b) Rubrospinal tract : There are several reports that section of the rubrospinal tract produces the slight deviation of the head for the short period chiefly to the opposite side or occasionally to the same side of the lesion (PIKE, ELABERG, McCULLOCH and CHAPPELL, 1931⁴⁴) ; KELLER and HARE, 1934³⁰) ; CARPENTER, 1956³⁶) ; ORIOLI and METTLER, 1956³⁸). However, in the present study, there was no case in which only the rubrospinal tract had been destroyed.

(c) Interstitial tract : HASSLER and HESS (1954)³⁰) demonstrated that stimulation and destruction of the vicinity of the interstitial nucleus and its tract may produce a cephalic deviation. However, in the present study, no case had the destruction of this system.

(d) Central tegmental tract : INGRAM, RANSON and their associates (1932)²⁸) evoked cephalic movements toward the opposite side by faradic stimulation of the central

tegmental tract at its rostral level. BEBIN (1956)²⁾ reported that stimulation of the central tegmental tract caused raising and deviation of the uvula, contraction of the palatal muscles and jaw, and movement of the shoulder mainly on the same side of the stimulation, or occasionally on both sides in monkey. However, in the present study, the role of the central tegmental tract on production of cephalic deviation was not confirmed, for in some cases, in which spasmodic torticollis-like posture was not produced, the central tegmental tract had been destroyed, and in other cases, in which spasmodic torticollis-like posture was present, there was no lesion of the central tegmental at all.

Above-mentioned results indicate that the red nucleus, the rubrospinal tract and the central tegmental tract may not play the essential roles for production of cephalic deviation and even if cephalic deviation is accompanied with a lesion of one or two of these regions, such a lesion seems to be secondary or accidental.

Accordingly, it is apparent that the combined lesions of the medial longitudinal fasciculus, the decussation of the brachium conjunctivum and the medial reticular formation, especially the reticular formation surrounding the dorsomedial part of the red nucleus at the level of the magnocellular part of the red nucleus cause the production of spasmodic torticollis-like posture. The present experimental data also suggest that, among these structures, the extent of the damage of the reticular formation surrounding the dorsomedial part of the red nucleus has the most important relation to the severity of spasmodic torticollis-like posture.

EXPERIMENT III

E. M. G. CHARACTERISTICS AND MECHANISM OF EXPERIMENTAL SPASMODIC TORTICOLLIS-LIKE POSTURE

This experiment is designed to study E. M. G. discharges and mechanism of spasmodic torticollis-like posture which was produced in Experiment II.

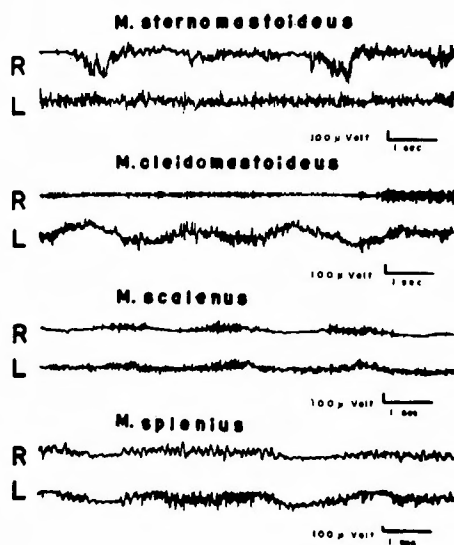
1. *E. M. G. Findings*

Electromyographic studies of spasmodic torticollis-like posture were carried out on 5 cats with marked torticollis, whose mesencephalic tegmentum had been destroyed for 5 to 7 days. Within these periods, all other neurological symptoms had disappeared almost completely and only spasmodic torticollis-like posture persisted.

All muscles of the neck were exposed under Nembutal anesthesia (25-35 mg/kg, intraperitoneal injection). Bipolar needle electrodes were inserted into Mm. sternocleidomastoideus, scalenus, splenius, levator scapulae, complexus, biventer and obliquus inferior, all of which seemed to play a role for deviation of the head. After recovery from the anesthesia, electromyograms were picked up from these muscles without any restraint of the movements of the whole body. There were no uniform patterns of electromyographic findings at rest in cases with spasmodic torticollis-like posture. Continuous or irregular discharges were recorded from all the muscles of the neck on both sides, but these discharges were usually more marked on the side of the mesencephalic lesion, i. e. on the side of the direction of the chin, as shown in Fig. 4.

It is generally accepted that torticollis is due to hyperactivity of the neck muscles chiefly on the same side of the deviated occiput. However, in our experiments chin of

E. M. G. (No. 110)



E. M. G. (No. 110)

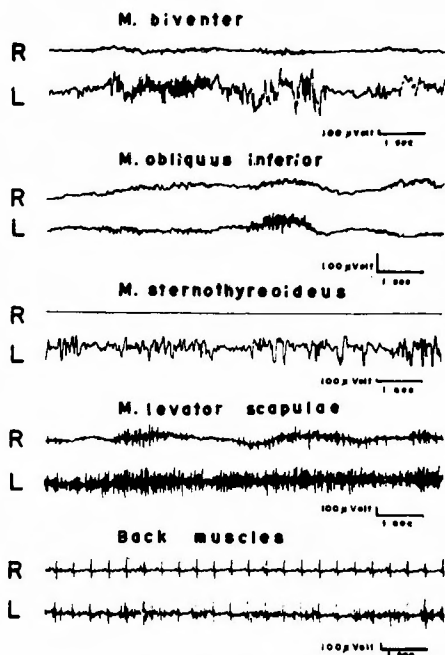


Fig. 4 Spasmodic torticollis-like posture with deviated chin toward the left was produced by the destruction of the left mesencephalic tegmentum.

the animals with spasmodic torticollis-like posture was deviated toward the side of the neck muscles showing more marked discharges. Thus, the following experiments were carried out in order to know this seemingly paradoxical mechanism.

2. Effects of mesencephalic stimulation upon alpha motoneurons in the 3rd cervical and accessory nerves

Bipolar electrodes were inserted stereotaxically into the same portions of the mesencephalic tegmentum as those of the lesion essential to the production of spasmodic torticollis-like posture, and the effects of the mesencephalic stimulation upon cervical and accessory alpha motoneurons were tested.

The electrodes used for the mesencephalic stimulation consisted of steel wire, being about 0.3 mm in diameter, and insulated except for a bare tip. Laminectomy of the upper cervical segments was performed under Nembutal anesthesia (25 mg/kg, intravenous injection) to expose the bilateral 2nd to 4th cervical roots. And the 3rd cervical dorsal roots on both sides were ligated at the distal part, and then placed on bipolar electrodes of silver wire for stimulation. The bilateral 3rd cervical and spinal accessory nerves were exposed near their innervated muscles, and severed at the peripheral ends. The severed ends of the nerves were placed on the other bipolar silver wire electrodes for picking-up the efferent outflows. A CR amplifier (time constant : 0.5 sec.) feeding a cathode-ray oscilloscope was used for recording the efferent outflows from the peripheral nerves. Single rectangular current pulses of 1.0 msec. in duration, 3-6 V in strength, were applied as stimuli to the mesencephalic tegmentum.

Fig. 5a

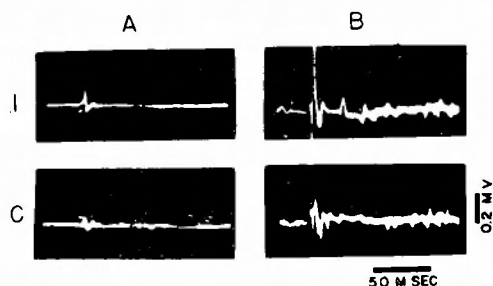
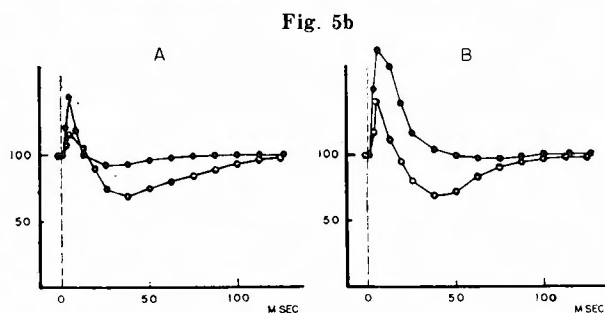


Fig. 5 Fig. 5a Efferent discharges recorded from the spinal accessory nerves (A) and the 3rd cervical nerves (B) by the stimulation of the mesencephalic tegmentum. I : Recorded from the ipsilateral nerves. C : Recorded from the contralateral nerves.

Fig. 5b Effects of the mesencephalic stimulation upon the mono-synaptic reflexes of the accessory nerves (A) and the 3rd cervical nerves (B). Ordinate : Reflex amplitudes recorded from the peripheral nerves in percentage of the control reflex size. Abscissae : Time interval in millisecond between conditioning and test stimuli. Open circles : Reflexes recorded from the contralateral nerves. Filled circles : Reflexes recorded from the ipsilateral nerves.



By the mesencephalic stimulation, marked discharges were recorded from both cervical and accessory nerves of bilateral sides (Fig. 5a), whereas discharges from the 3rd cervical nerves were usually more marked than those from the accessory nerve. And they appeared more dominantly on the ipsilateral side than the contralateral one. Maintained discharges without any electrical stimulation were often recorded from the 3rd cervical nerves of bilateral sides, while such discharges were hardly seen from the accessory nerves on either side. This fact may indicate that the neck muscles connected with the 3rd cervical nerve are tonic in nature while those with the accessory nerve are rather phasic.

After recording the efferent outflows to the peripheral nerves elicited by the mesencephalic stimulation, influences of the stimulation upon the monosynaptic reflex in the accessory and the 3rd cervical nerves were tested. The monosynaptic reflex discharges elicited by the stimulation of the 3rd cervical dorsal root were recorded from both the 3rd cervical and accessory nerves, and conditioned by the mesencephalic stimulation with varying time intervals. Fig. 5b illustrates the time course of facilitatory and inhibitory influences of stimulating the mesencephalic tegmentum on the monosynaptic spinal reflexes. As seen in the figure, the mesencephalic stimulation facilitated the monosynaptic reflex recorded from the ipsilateral 3rd cervical nerves at time interval from a few msec. to about 50 msec., while it facilitated slightly and then inhibited the monosynaptic reflex recorded from the contralateral 3rd cervical nerve at a time interval from about 15 to 100 msec. The mesencephalic stimulation also facilitated the monosynaptic reflex recorded from the ipsilateral accessory nerve at time interval from about 3 to 12 msec., while it facilitated slightly and then inhibited the monosynaptic reflex recorded from the contralateral one at time interval from about 15 to 100 msec. or more.

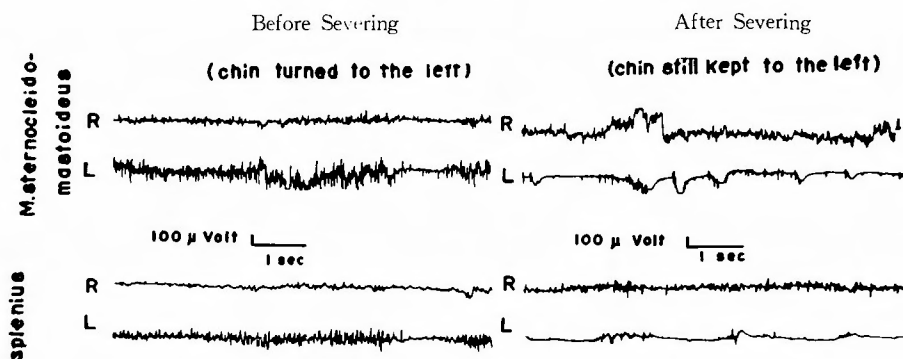
Accordingly, it is assumable that the 3rd cervical nerve may participate in innervation of tonic activity of the neck muscles rather than phasic one, because the 3rd cervical nerve showed often the maintained discharges and was facilitated by the mesencephalic stimula-

tion for a relatively long time when compared with the accessory nerve. On the other hand, the accessory nerve seems to participate in innervation of the phasic activity of the neck muscles rather than the tonic one, for the accessory nerve revealed scarcely maintained discharges and was facilitated with a short time course by the mesencephalic stimulation.

Consequently, the following assumption may be given. The 3rd cervical and accessory nerves keep balance in normal condition under the influences of the facilitation from the ipsilateral upper centers including the mesencephalic tegmentum and of the inhibition from the contralateral ones. When a lesion is made in the mesencephalic tegmentum, the chin must be slightly rotated around the longitudinal axis and markedly around the vertebral axis toward the side of the lesion, and slightly overextended backwards by the combined activities of the neck muscles innervated chiefly by the 3rd cervical nerve, such as *Mm. longissimus capitis et cervicis*, *interspinalis cervicis*, *multifidus cervicis*, *longus cervicis et capitis* and *iliocostalis cervicis*. And when emotional excitations are induced by any means, the accessory nerve gets into action, resulting in spasmodic movements of the head, for this nerve innervates chiefly *Mm. sternocleidomastoideus* and *trapezius*.

3. Effect of severance of the upper cervical dorsal roots upon the muscle discharges

The 2nd to 5th cervical dorsal roots were severed on the side of the mesencephalic lesion, which had been made 7 days before. E. M. G. discharges were greatly decreased by this operation, changing the pattern into the bursts of the short duration (Fig. 6). However, rotation of the head with deviated chin toward the side of the lesion still remained. Therefore, the marked continuous discharges of the neck muscles on the same side seem to be induced reflexly by the proprioceptive afferent impingements from the muscles. In other cases, the upper cervical dorsal roots of the same segments were severed on the opposite side. The chin was rotated toward the side of the severance of the cervical dorsal roots, i. e. toward the side opposite the mesencephalic lesion. This fact indicates that spasmodic torticollis-like posture of cat is also effected by hyperactivity of the gamma motor system of the side opposite the mesencephalic lesion, for severance of the dorsal roots may eliminate the effect of gamma motor system by disappearance of the afferent impulses from the muscles spindles. It may be thus, postulated that cephalic de-



Spasmodic torticollis-like posture was produced by electrolytic lesions of the left brain stem in this cats (No. 154).

Fig. 6 E. M. G. Before and After Severing The Left Cervical Posterior Roots (C_2, C_3, C_4, C_5),

viation with rotated chin toward the side of the mesencephalic lesion may be due to a more intense contraction of the neck muscles on the opposite side than that on the ipsilateral one.

E. M. G. findings showed that discharges were more marked on the same side than on the opposite one, though the chin of cat was turned toward the side of the lesion. This finding does not seem to agree with the general opinion that deviation of the head toward the one side may be due to hyperactivity of the neck muscles on the other side.

Further studies in this regard be performed to elucidate mechanism of the revelation of the paradoxical E. M. G. discharges.

SUMMARY

1. Experimental spasmodic torticollis-like posture was produced in 80 (marked one in 53, moderate one in 14 and slight one in 13) out of 102 cats by unilateral destruction of the mesencephalic tegmentum. The etiologic lesion encroached upon the portions of the decussation of the brachium conjunctivum, the medial longitudinal fasciculus and the medial reticular formation, especially the reticular formation surrounding the dorsomedial part of the red nucleus at the level of the magnocellular part of the red nucleus.

2. The chin was deviated toward the same side of the mesencephalic lesion, usually accentuated by emotional stress.

3. In E. M. G. findings, irregular muscle discharges from the various neck muscles were usually more marked on the same side of the mesencephalic lesion than those on the opposite side. It was unable to find out the relationship between the patterns of the muscle discharges and the movements and or posture of the head.

4. Effect of mesencephalic stimulation upon alpha motoneurons in the 3rd cervical and accessory nerves was studied. The mesencephalic stimulation facilitated the reflex firing of motoneurons in the ipsilateral 3rd cervical and accessory nerves and inhibited the reflex firing of motoneurons in the contralateral 3rd cervical and accessory nerves.

5. It was noted, by severing the cervical dorsal roots, that the cephalic deviation was also due to unbalanced activities of the gamma motor system which were induced by the mesencephalic lesion.

PART II

THE EFFECTS OF DESTRUCTION OF THE BASAL GANGLIA UPON THE SPASMODIC TORTICOLLIS-LIKE POSTURE

In Part I spasmodic torticollis-like posture was produced by destruction of the portions of the medial longitudinal fasciculus, the decussation of the brachium conjunctivum and the medial reticular formation at the level of the magnocellular part of the red nucleus, especially the reticular formation surrounding the dorsomedial part of the red nucleus. Although spasmodic torticollis-like posture in human cases can be divided into several types, it is true that some cases of the human spasmodic torticollis are quite similar in nature and mechanism as that of experimental spasmodic torticollis. Thus, we may consider that if such an experimental spasmodic torticollis which was produced by the mesencephalic

lesion can be cured by a lesion in some portions of the basal ganglia, good results may also be expected in human cases with spasmodic torticollis by stereotaxic surgery which aims to destroy or inactivate the basal ganglia.

Under the above-mentioned idea, the following experiment was carried out on (1) whether such an abnormal posture of the head is improved by destruction of the selected portions of the basal ganglia and (2) destruction of which nuclei of the basal ganglia and how extensive lesions are the most effective.

1. *Material and Method*

Thirty seven selected cats with marked spasmodic torticollis-like posture were used in this experiment, cats with the following conditions were excluded : animals (1) being crestfallen and/or enfeeble, (2) of slight or moderate spasmodic torticollis-like posture, (3) with any infection, (4) with severe additional neurological deficits, such as pronounced motor disturbances, ataxia, etc.

All the animals were anesthetized with Nembutal (25~35 mg/kg, intraperitoneal injection) and then operated under sterile condition by stereotaxic technique to insert an electrode, which is the same electrode used for giving a damage to the mesencephalic tegmentum, into the basal ganglia. Electrolytic lesion was made with a direct current of 3 MA, 10~20 V, for 15~25 sec. through an inserted positive monopolar electrode into the following locations according to the classification of WALKER : (1) the posterior basal part of the ventrolateral nucleus of the thalamus (VL), (2) the anterior basal part of VL, (3) the medial part of VL, (4) the zona incerta including FOREL'S field H_1 and H_2 , and (5) the nucleus entopeduncularis of the pallidum, all of which are considered to be somewhat concerned with the mechanism of spasmodic torticollis-like posture.

After the observation for the period of several weeks, the cats were sacrificed. The brains were perfused *in situ* with 200~500 ml of 10% formalin solution, and removed carefully. Blocks of all the brains for microscopic study were embedded in celloidin, sectioned serially at the thickness of 20 microns and stained with the modified KLÜVER-BARRERA'S method to find the location of the lesion in the basal ganglia.

2. *Results*

(1) The posterior basal part of VL

This region is roughly equal to V.o.p., which was denominated by HASSLER. One of the most complete investigations of the nuclear configuration, connections and functions of the thalamus was made by HASSLER (1950¹⁸⁾, 1955¹⁹⁾), based upon study of human materials.

- a) Destruction of the side of the mesencephalic lesion (i. e. the same side of the direction of the rotated chin) (2 cases)

All of the 2 cats showed no spasmodic torticollis-like posture.

- b) Destruction of the side opposite the mesencephalic lesion (i. e. the side opposite the direction of the rotated chin) (5 cases)

Two out of 5 cats showed improvement immediately after the destruction (Fig. 7) (1 : slightly, 1 : markedly), although cerebellar symptoms, such as motor disturbances and ataxia, persisted until the time of sacrifice. The remaining 3 showed neither improvement nor deterioration in any degree.



Fig. 7 a



Fig. 7 b

Fig. 7 Cat No. 82 Eig. 7 a shows the torticollis on the 4th day after destruction of the left mesencephalic tegmentum. The posterior basal part of VL on the right was destroyed on the next day. Fig. 7 b shows the posture of the head on the 3rd day after treatment.

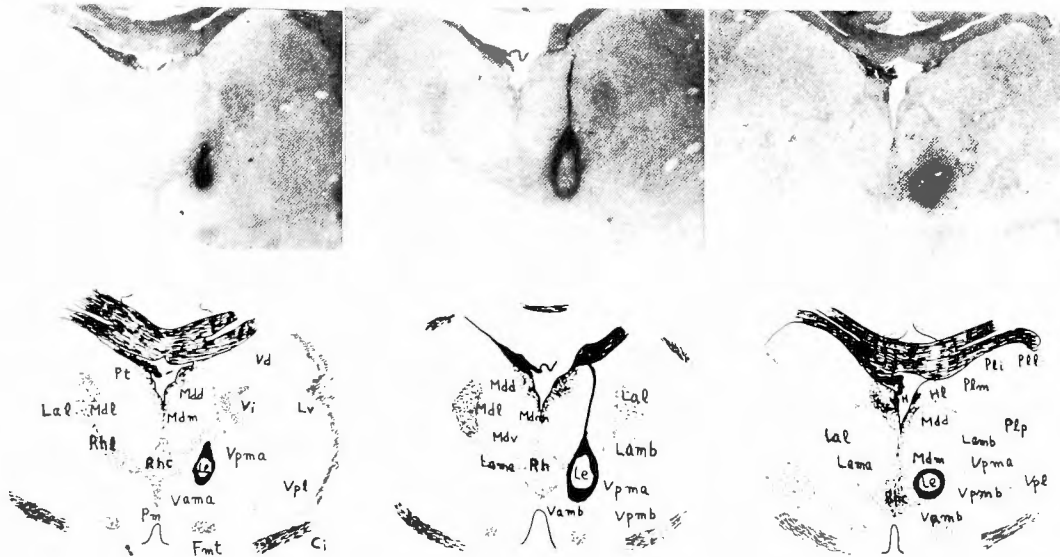


Fig. 8 Photomicrographs of cross section of the cerebrum in cat No. 78

Histological findings (Table 6; Fig. 8): According to the classification of HIROKAWA (1941)²¹⁾, the lesion common to 2 cats (No. 78 and 82), in which improvement was observed, had infringed on the almost whole part of pars anterior medialis a and b of the nucleus ventralis (Vama and Vamb), the rostral part of pars posterior medialis a and b of the ventral nucleus (Vpma and Vpmb) and portions of the medial, median and posterior nuclear groups. In remaining 3 (No. 83, 84 and 97), in which no improvement was seen, the lesions had encroached more caudally, and the portion of Vama and Vamb, and the rostral part of Vpma were intact.

Table 6

Destruction of the posterior basal part of VL

	improved		not improved		
	78	82	83	84	97
Val					
Vama	0	0			
Vamb	0	0		0	0
Vpma	0	0	0	0	0
Vpmb		0	0		
Vpl			0	0	0
Vi			0		0
Rt					
medial nuclear group	Mdv Mdd Mv Pf	Mdv Lamb Mdl Lal	Lam	Lama Lamb	Lamb
median nuclear group		Rad Rav			
posterior nuclear group	Plm Plsp Li				

Table 7

Destruction of the anterior basal part of VL

	improved				not improved
	79	85	87	112	80
Val	0	0		0	0
Vama	0	0	0	0	0
Vamb	0		0	0	
Vpma	0	0	0	0	
Vpmb	0		0		
Vpl		0	0	0	
Vi	0	0	0	0	0
Rt					
anterior nuclear group	Amv Av	Amv		Amd Av	
medial nuclear group	Md	Lal		Lam Lal	
lateral nuclear group	Lda Lv	Lv			

(2) The anterior basal part of VL

This region is approximately equal to V.o.a., which was denominated by HASSLER.

a) destruction of the side of the mesencephalic lesion (2 cases)

All of the 2 cats showed neither any discernible improvement nor deterioration.

b) destruction of the side opposite the mesencephalic lesion (5 cases)

Four out of 5 cats showed improvement of spasmodic torticollis-like posture immediately after operation (Fig. 9) (2 : markedly, 2 : moderately). Improvement persisted without any deterioration for several weeks until animals were sacrificed. Additional neurological deficits were not accompanied. The remaining one was found to have neither improvement nor deterioration.

Histological findings (Table 7; Fig. 10) : The lesion common to 4 cats (No. 79, 85, 87 and 112), in which improvement was observed, had infringed on almost whole part of Vama, the rostral part of Vpma and part of pars intermedia (Vi.) In the remain-

ning one (No. 80), in which improvement was not seen, the lesion had infringed more rostrally, and Vpma was not destroyed.

(3) The medial part of VL

This region is roughly equal to V.o.i., which was denominated by HASSLER.

a) Destruction of the side of the mesencephalic lesion (2 cases)

One of 2 cats showed slight improvement. The other one showed neither improvement nor deterioration.

Histological findings: The lesion of cat No. 93 with slight improvement had encroached upon Vama and Vpma.

b) Destruction of the side opposite the mesencephalic lesion (4 cases)

Three out of 4 cats showed improvement immediately after operation (Fig. 11) (2: markedly, 1: moderately). Improvement persisted without any deterioration for several weeks. The remaining one was found to have neither any discernible improvement nor deterioration.

Histological findings (Table 8; Fig. 12): The lesion common to 3 cats (No. 62, 64 and 92) with improvement had infringed mainly on the portions of Vama, Vamb, Vpma, Vpmb and pars posterior lateralis (Vpl). In the remaining one (No. 99), in which no improvement was observed, the lesion had encroached more medially and the portion of the medial and lateral nuclear groups had been destroyed. However, the portion of the nucleus ventralis was intact.

(4) Region of the zona incerta

This region includes the zona incerta, FOREL'S field H_1 and H_2 , and the portion of the posterior basal part of VL.

a) Destruction of the side of the mesencephalic lesion (2 cases)

All of the 2 cats were found to have neither improvement nor deterioration.

b) Destruction of the side opposite the mesencephalic lesion (6 cases)

Four out of 6 cats showed improvement immediately after operation (Fig. 13) (3: markedly, 1: moderately). On the 3rd or 4th day postoperatively, severe neurological



Fig. 11 a



Fig 11 b

Fig. 11 Cat No. 62 Fig. 11 a shows the torticollis on the 6th day after destruction of the left mesencephalic tegmentum. The medial part of VL on the right was destroyed on that day. Fig. 11 b shows the posture of the head on the 4th day after treatment.



Fig. 13 a



Fig. 13 b

Fig. 13 Cat No. 90 Fig. 13 a shows the torticollis on the 5th day after destruction of the left mesencephalic tegmentum. The portion of the zona incerta on the right was destroyed on the next day. Fig. 13 b shows the posture of the head on the 5 day after treatment.

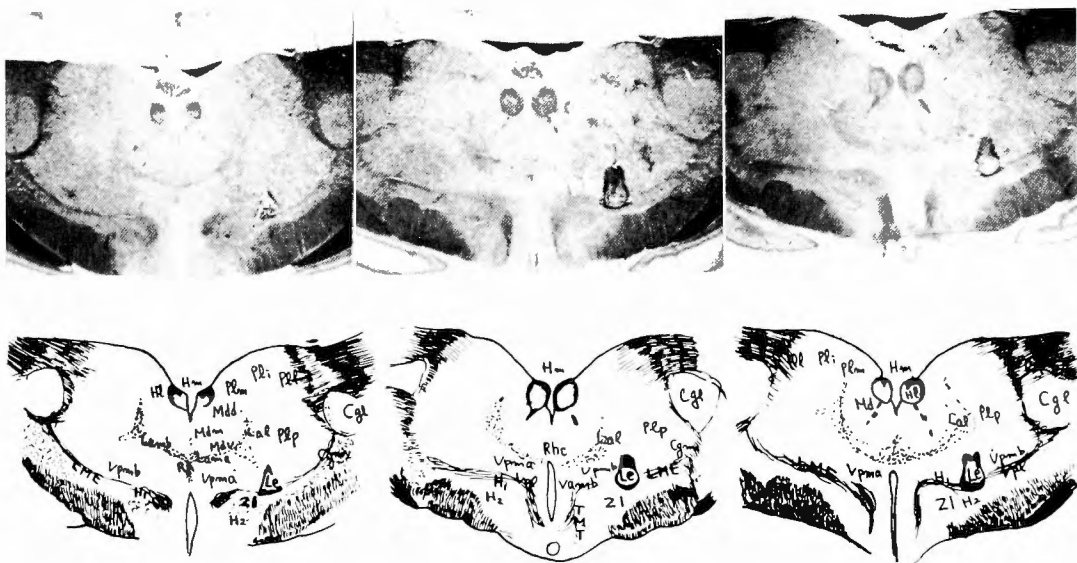


Fig. 14 Photomicrographs of cross section of the cerebrum in cat No. 68

deficits appeared, such as marked disturbances of gait, peduncular movement of the head and ataxia, and cats usually lowered themselves and staggered with over-steps and were unable to jump.

Histological findings (Table 9, Fig. 14) : The lesion common to 4 cats (No. 68, 81, 90 and 118) with improvement had infringed on the portions of Vpma, Vpl, the zona incerta, the lamina medullaris externa and FOREL's field H₁ and H₂. In the remaining 2 (No. 76 and 96), in which no improvement was observed, the lesions had encroached upon the portions of Vpl, the zona incerta, the lamina medullaris externa and the nucleus reticularis, but no lesion upon the portions of Vpma and FOREL's field H₁ and H₂.

(5) The nucleus entopeduncularis of the pallidum

This nucleus is said to be roughly equal to the medial segment of the pallidum in human, although this nucleus does not belong to the latter.

a) Destruction of the side of the mesencephalic lesion (2 cases)

All of the 2 cats were found to have neither improvement nor deterioration.

b) Destruction of the side opposite the mesencephalic lesion (7 cases)

Five out of 7 cats showed improvement immediately after operation (Fig. 15) (3 : markedly, 2 : moderately). In all cases which had marked improvement, additional neurological deficits were not seen. Improvement persisted for whole duration of the observation (several weeks).

Histological findings (Table 10 ; Fig. 16) : The lesion common to 5 cats (No. 56, 89, 100, 103 and 114) with improvement had infringed on a half or more part of the nucleus entopeduncularis. In 4 of them (No. 56, 100, 103 and 114), the lesion had encroached upon the portion of the ansa lenticularis. In remaining 2 (No. 98 and 102)



Fig. 15 a



Fig. 15 b

Fig. 15 Cat No. 56 Fig. 15 a shows the torticollis on the 10 th day after destruction of the left mesencephalic tegmentum. The right nucleus entopeduncularis was destroyed on that day. Fig. 15 b shows the posture of the head on the 3rd day after treatment.

Table 10
Destruction of the pallidum

	improved					not improved	
	56	89	100	103	114	98	102
nucleus entopeduncularis	$\frac{1}{2}$	$\frac{1}{1}$	$< \frac{1}{2}$	$\frac{1}{2}$	$< \frac{1}{2}$	$\frac{1}{3}$	$< \frac{1}{2}$
internal capsule	0				0	0	0
optic tract	0			0			
ansa lenticularis	0		0	0		0	

< : more than > : less than



Fig. 16 Photomicrographs of cross section of the cerebrum in cat No. 89

which had no improvement, the lesion had encroached less extensively than that of the former.

As above-mentioned, 19 out of 37 cases had improvement after the destruction either of thalamic or pallidal lesion was made. Out of 19, 10 cases had a lesion in the thalamus, 4 in the region of the zona incerta, and 5 in the pallidum, in all of which except one case with the thalamic lesion, the destruction was made on the side opposite the mesencephalic lesion (Table 11). In 9 cases which had a lesion in the thalamus on the side opposite the mesencephalic lesion, Vama and Vpma were destroyed together in all these 9 cases (100%), Vamb in 8 (89%), Vpmb, Vpl and Vi in 6 (67%) and Val in 5 (56%) (Table 12). Thus, if a lesion is made in the portion extending over the rostral half of Vpma and the caudal part of Vama on the side opposite the mesencephalic lesion, spasmodic torticollis-like posture may be considered to be cured. Regarding a lesion of the zona incerta, all cases were usually accompanied with cerebellar deficits, even if marked improvement was observed for spasmodic torticollis-like posture. Thus, the region of the

Table 11
Effects of destruction of the basal ganglia

	contralateral		ipsilateral	
	improved	not improved	improved	not improved
posterior basal part of VL	2	3	0	2
anterior basal part of VL	4	1	0	2
medial part of VL	3	1	1?	1
zona incerta	4	2	0	2
nucleus entopeduncularis	5	2	0	2
Total	18	9	1?	9

Table 12
Destruction of the nucleus ventralis of the thalamus

	improved torticollis No. of animals	%
pars anterior lateralis (Val)	5	56
pars anterior medialis (Vam)		
dorsolateral part (Vama)	9	100
ventromedial part (Vamb)	8	89
pars posterior medialis (Vpm)		
dorsomedial part (Vpma)	9	100
ventrolateral part (Vpmb)	6	67
pars posterior lateralis (Vpl)	6	67
pars intermedia (Vi)	6	67

zona incerta seems not to be selected as a target point for the treatment of spasmodic torticollis-like posture. Regarding the pallidum, more extensive lesion in the pallidum seems to be effective for the treatment of spasmodic torticollis-like posture.

From above-mentioned results, it seems that the following locations should be selected as target points for the treatment of experimental spasmodic torticollis-like posture : (1) the portion extending over the rostral half of Vpma and the caudal part of Vama in the thalamus or (2) the nucleus entopeduncularis of the pallidum on the side opposite the mesencephalic lesion.

3. Stimulation of the basal ganglia

As shown in above-mentioned results, the portion extending over the rostral half of Vpma and caudal part of Vama, and the nucleus entopeduncularis seem to be target points for the treatment of spasmodic torticollis-like posture. Thus, the following experiments were carried out in order to know the relationship between these areas and the mesencephalic tegmentum. A brief rectangular current pulses lasting 3 to 5 seconds (2~3 msec. of pulse width, 2~8 V, 8~12 cycle/sec. in frequency ; or 1~3 msec. of pulse width, 2~5 V, 60~100 cycle/sec. in frequency) were applied as stimuli through implanted bipolar electrodes.

- i) Portion extending over the rostral half of Vpma and the caudal part of Vama (9 cases)

In all of the 9 cats (2 : intact, 7 : with marked spasmodic torticollis-like posture),

the chin was rotated toward the opposite side of the stimulation by the stimulation either of low or high frequency. In cases with the marked spasmodic torticollis-like posture, rotation of the head was extremely accentuated by the stimulation of this portion on the opposite side of the mesencephalic lesion, and became un conspicuous and occasionally returned normal position by the stimulation on the same side. Other symptoms were usually accompanied, such as increasing of the muscles tone of extremities, slow-moving (or occasionally stopping of movement), falling to the side of stimulation, extending and raising of the forelimb on the opposite side, and, occasionally, tremor of the same forelimb.

On the other hand, cephalic deviation was not induced by the stimulation of the various other parts of the thalamus, such as the medial part of the nucleus posterior (2 cases), the dorsolateral part of the nucleus ventralis postero-medialis (2 cases), the nucleus centrum median (2 cases), the nucleus ventralis anterior (2 cases), the ventromedial part of the nucleus lateralis posterior (2 cases), the nucleus anterior medialis (2 cases), the nucleus medialis dorsalis (1 case), the nucleus centralis lateralis (1 case) and the nucleus ventralis posterolateralis (1 case).

ii) Region of the zona incerta (5 cases)

In all of the 5 cats (2 : intact, 3 : with marked spasmodic torticollis-like posture), the chin was rotated toward the opposite side of the stimulation by the stimulation either of low or high frequency. Other symptoms were usually induced, such as increasing of the muscle tone, hypo- or akinesia, chewing movement, and occasionally, extending and/or scratching movements of the forelimb on the opposite side. If stimuli were intensified, convulsion was induced on the contralateral side of the body in 4 cats.

iii) Nucleus entopeduncularis (5 cases)

In all of the 5 cats (2 : intact, 3 : with marked spasmodic torticollis-like posture), the chin was rotated toward the opposite side of the stimulation by the stimulation either of low or high frequency. Increasing of the muscles tone, curvature of the body toward the opposite side, and, occasionally, chewing movement and sniffing were also noted.

These studies also indicate that destruction either of the portion extending over the rostral half part of Vpma and the caudal part of Vama in the thalamus or of the nucleus entopeduncularis of the pallidum may alleviate spasmodic torticollis-like posture which was produced by destruction of the mesencephalic tegmentum.

4. *Comment*

With the introduction of stereotaxic surgery, excellent results have been obtained for the treatment of many kinds of abnormal involuntary movements, such as parkinsonism, athetosis, cerebral palsy, etc. However, it has not yet been generally believed that spasmodic torticollis may be alleviated by stereotaxic surgery. Even the procedure of reducing the strength of the participating neck muscles has not always produced a satisfactory result.

In the present study, it was confirmed that the lesion either of the portion extending over the rostral part of Vpma and the caudal part of Vama or of the nucleus entopeduncularis may alleviate spasmodic torticollis-like posture which was produced by destruction of the mesencephalic tegmentum. Therefore, it seems to be necessary to survey the relationship between these above-mentioned locations and the mesencephalic tegmentum.

1) Portion extending over the rostral part of Vpma and the caudal part of Vama

This region is roughly equal to the portion posterior to the anterior basal part of VL (WALKER), to the slightly caudal part of V.o.i. (HASSLER) and to the portion extending over vtm and vb (RIOCH).

It is well known that many fibers enter the thalamic nucleus from the ipsilateral tegmentum (CARPENTER, 1956³⁾; CARPENTER and STEVENS, 1957⁹⁾; JONSON and CLEMENTE, 1959²⁹⁾. HASSLER (1949)¹⁷⁾ and HASSLER and HESS (1954)²⁰⁾ pointed out that V.o.i. contains not only few fibres of the brachium conjunctivum but also a large contingent from the nucleus interstitialis. Thus, it may be postulated that destruction of V.o.i. may reduce the unbalanced activities which produced by the unilateral mesencephalic lesion.

2) Nucleus entopeduncularis of the pallidum

The globus pallidus sends fibers directly to the mesencephalic tegmentum and the prerubral field of the ventral thalamic nucleus and through synaptic connections, in the ventral thalamus by way of the ansa lenticularis and the lenticular fasciculus (PAPEZ, 1942⁴⁴⁾; WOODBURN, CROSBY and MCCOTTER, 1946⁵³⁾; JONSON and CLEMENTE, 1959²⁹⁾. A few fibers also enter the medial segment of the globus pallidus from the mesencephalic tegmentum. Moreover, it has been well known that destruction of the pallidum reduces rigidity on the opposite side. Thus, the destruction of the globus pallidus may alleviate spasmodic torticollis-like posture by anatomical relation to the mesencephalic tegmentum and also by reducing rigidity.

From above-mentioned account, it may be considered that even destruction of the contralateral mesencephalic tegmentum may alleviate spasmodic torticollis-like posture. Thereupon, in 3 cats in which spasmodic torticollis-like posture was produced by destruction of the mesencephalic tegmentum, the other side was destroyed in the same degree. The position of the chin was also returned to normal position. However, additional severe neurological deficits, such as staggering gait, disturbances of equilibrium, hypotonia of both forelimbs, disturbances of chewing movement, etc., were accompanied in all 3 cases, and were more severe than those induced by destruction of the region of the zona incerta. Such symptoms improved very slowly, persisting for several weeks.

Thereupon, spasmodic torticollis-like posture produced experimentally may be alleviated by destruction or inactivation of the basal ganglia, or the contralateral mesencephalic tegmentum, reducing the unbalanced activities.

SUMMARY

1. Spasmodic torticollis-like posture in Part I was able to be improved by destruction of the selected portions of the basal ganglia. These were (a) the portion extending over the rostral half of pars posterior medialis a and the caudal part of pars anterior medialis a of the nucleus ventralis of the thalamus (=slightly caudal part of V.o.i.) and (b) the nucleus entopeduncularis (=the medial segment of the pallidum).

2. Such an alleviation is due to the reduction of the unbalanced activities by the anatomical and physiological relation.

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REFERENCES

- 1) Alpers, B. J., and C. S. Drayers : The organic back ground of some cases of spasmodic torticollis. Report of a case with autopsy. *Am. J. med. Sci.*, **1937**, 378-384, 1937.
- 2) BeBin, J. : The central tegmental bundle. *J. comp. Neurol.*, **105**, 287-332, 1956.
- 3) Carpenter, M. B. : A study of the red nucleus in the rhesus monkey. *Ibid.*, **105**, 195-250, 1956.
- 4) Carpenter, M. B., G. M. Brittin and J. Pines . Isolated lesions of the fastigial nuclei. *Ibid.*, **109**, 65-89, 1958.
- 5) Carpenter, M. B., and G. H. Stevens : Structural and functional relationships between the deep cerebellar nuclei and the brachium conjunctivum in the rhesus monkey. *Ibid.*, **107**, 109-163, 1957.
- 6) Carrea, R. M. E., and F. A. Mettler : Function of the primate brachium conjunctivum and related structures. *Ibid.*, **102**, 151-332, 1955.
- 7) Cassierer, R. : Halsmuskelkrampf und Torsionsspasmus. *Klin. Wschr.*, **1**, 53-57, 1922.
- 8) Cohen, D., W. W. Chambers and J. M. Sprague : Experimental study of the efferent projections from the cerebellar nuclei to the brain stem of the cat. *J. comp. Neurol.*, **109**, 233-260, 1958.
- 9) Dow, R. C. : Effects of unilateral and bilateral labyrinthectomy in monkey, baboon and chimpanzee. *Am. J. Physiol.*, **121**, 392-399, 1938.
- 10) v. Economo, C. J., and J. P. Karplus : Zur Physiologie und Anatomie des Mittelhirns. *Arch. Psychiat. Nervenkr.*, **46**, 275-356, 1909.
- 11) Ferraro, A., and S. E. Barrera : Effects of lesions of the juxtarestiform body (I. A. K. Bundle) in macacus rhesus monkeys. *Arch. Neurol. & Psychiat.*, **25**, 13-28, 1936.
- 12) Ferraro, A., B. L. Pacella, and S. E. Barrera : Effects of lesion of the medial vestibular nucleus. *J. comp. Neurol.*, **79**, 185-245, 1940.
- 13) Foerster, O. : Torticollis spasticus. *Ztschr. f. orthop. Chirurg.*, **51**, 144, 1929.
- 14) Foerster, O. : Mobile spasm of the neck muscles and its pathological basis. *J. comp. Neurol.*, **58**, 1043, 1933.
- 15) Foltz, E. L., L. M. Knopp, and A. A. Ward, Jr. : Experimental spasmodic torticollis. *J. Neurosurg.*, **16**, 55-72, 1959.
- 16) Grinker, R. R., and A. E. Walker : The pathology of spasmodic torticollis with a note on respiratory failure from anesthesia in chronic encephalitis. *J. Nerv. Ment. Dis.*, **78**, 630, 1933.
- 17) Hassler R. : Über die Rinden- und Stammhirnanteile des menschlichen Thalamus. *Psychiat. & Neurol.*, **1**, 181-187, 1949.
- 18) Hassler, R. : Anatomie des Thalamus. *Arch. f. Psychiat.*, **184**, 249-256, 1950.
- 19) Hassler, R. : Functional anatomy of the thalamus VI Congr. Latino-america, de Neurochiria Montevideo, 754-787, 1955.
- 20) Hassler, R., and W. R. Hess : Experimentelle und anatomische Befunde über die Drehbewegungen und ihre nervösen Apparate. *Arch. Psychiat.*, **192**, 448-526, 1954.
- 21) Herz, E., and G. H. Glaser : Spasmodic torticollis. 2. Clinical evaluation. *Arch. Neurol. & Psychiat.*, **61**, 129-136, 1949.
- 22) Herz, E., and P. F. Hoefel : Spasmodic torticollis. 1. Physiological analysis of involuntary motor activity. *Ibid.*, **61**, 129-136, 1949.
- 23) Hinman, A., and M. B. Carpenter : Efferent fiber projections of the red nucleus in the cat. *J. comp. Neurol.*, **113**, 61-82, 1959.
- 24) Hirokawa, S. : Über die Thalamuskerneder Katze. *Japanese J. med. Sci.*, part I anatomy, **9**, 119-147, 1941.
- 25) Hyde, J. E., and A. B. Slusher : Functional role of medial longitudinal fasciculus in evoked conjugate deviations in cats. *Am. J. Physiol.*, **200**, 919-922, 1961.
- 26) Hydman, O. R. : Torticollis spasticus. Suggested etiologic relation to the vestibular apparatus : report of case. *Arch. Otolarynh.*, Chicago, **29**, 927-938, 1939.
- 27) Ingram, W. R., S. W. Ranson, and R. W. Barris : The red nucleus. Its relation to postural tonus and righting reactions. *Arch. Neurol. & Psychiat.*, **31**, 768-786, 1934.

- 28) Ingram, W. R., S. W. Ranson, F. I. Hannett, F. R. Zeiss, and E. H. Trewilliger : Results of stimulation of the tegmentum with the Horsley-Clarke stereotaxic apparatus. *Arch. Neurol. Psychiat.*, **28**, 513-544, 1932.
- 29) Jonson, T. H., and C. D. Clemente : An experimental study of the fiber connections between the putamen, globus, ventral thalamus, and midbrain tegmentum in cat. *J. comp. Neurol.*, **113**, 83-101, 1959.
- 30) Keller, A. D., and W. K. Hare : The rubro-spinal tract in the monkey. Effects of experimental section, 1934.
- 31) Kemberling, S. R., H. W. Baird, and E. A. Spiegel : Experimental torticollis of rhombencephalic origin. *J. Neuropathol. exp. Neurol.*, **11**, 184-191, 1952.
- 32) McMasters, R. E., and G. V. Russel : Efferent pathways from the deep cerebellar nuclei of the cat. *J. comp. Neurol.*, **110**, 205-219, 1958.
- 33) Mettler, F. A. : Extensive unilateral cerebral removals in the primate : Physiologic effects and resultant degeneration. *J. comp. Neurol.*, **79**, 185-243, 1943.
- 34) Mettler, F. A. : Physiologic consequences and anatomic degeneration following lesions of the primate brainstem : Plantar and patellar reflexes. *Ibid.*, **80**, 69-148, 1944.
- 35) Mettler, F. A. : Deviation of the head. *Arch. Neurol. & Psychiat.*, **81**, 442-457, 1959.
- 36) Mussen, A. T. : Experimental investigations of the cerebellum. *Brain*, **50**, 313-349, 1927.
- 37) Northington, P., and S. E. Barrera : Effects of unilateral and bilateral section of 8th nerve. *Arch. Neurol. & Psychiat.*, **32**, 52-71, 1934.
- 38) Orioli, F. L., and F. A. Mettler : The rubrospinal tract in macaca mulatta. *J. comp. Neurol.*, **106**, 299-318, 1956.
- 39) Orioli, F. L., and F. A. Mettler : Descending limb of the brachium conjunctivum in macaca mulatta. *Ibid.*, **106**, 339-362, 1956.
- 40) Orioli, F. L., and F. A. Mettler : Effect of rubrospinal tract section on ataxia. *Ibid.*, **107**, 305-313, 1957.
- 41) Orioli, F. L., and F. A. Mettler : Consequences of section of the simian restiform body. *Ibid.*, **109**, 195-204, 1958.
- 42) Papez, J. M. : A summary of fiber connections of basal ganglia with each other and with other portions of the brain. *Res. Pub. Assoc. Ros. Nerv. Menf. Dis.*, **21**, 21-68.
- 43) Patterson, R. M., and S. C. Little : Spasmodic torticollis. *J. Nerv. ment. Dis.*, **98**, 571-599, 1943.
- 44) Pike, F. H., C. A. Elsberg, W. S. McCulloch, and M. N. Chapell : Some observations on experimentally produced convulsions. II. The type of convulsions elicitable after lesions of the rubrospinal system, with some incidental findings. *Am. J. Psychiat.*, **10**, 567-594, 1931.
- 45) Powell, T. P. S., and W. M. Cowan : A study of thalamostriate relations in the monkey. *Brain*, **79**, 364-390, 1956.
- 46) Putnam, T. J., E. Herz, and G. H. Glaser : Spasmodic torticollis. 3. Surgical treatment. *Arch. Neurol. & Psychiat.*, **61**, 240-247, 1949.
- 47) Redemaker, G. G. J. : Die Bedeutung der roten Kerne und des übrigen Mittelhirns für Muskeltonus, Körperstellung und Labyrinthreflexe, Berlin Springer-Verlag, 1926.
- 48) Spiegel, E. A. : Der Tonus der Skelettmuskulatur 2nd. Berlin Springer, 1927.
- 49) Spiegel, E. A., and W. J. Bernis : Zentrum der statischer Innervation. *Arch. Neurol. Inst. Univ. Wien.*, **27**, 197, 1925.
- 50) Spiegel, E. A., M. S. Kletzkyn, E. G. Szekely, and H. T. Wycis : Role of Hypothalamic Mechanisms in Thalamic Pain. *Neurology*, **4**, 739-751, 1954.
- 51) Sprague, J. M., and W. W. Chambers : Regulation of posture in intact and decerebrate cat. 1. Cerebellum, reticular formation, vestibular nuclei. *J. Neurophysiol.*, **16**, 451-463, 1953.
- 52) Ward, A. A., Jr. : Decerebrate rigidity. *J. Neurophysiol.*, **10**, 89, 1947.
- 53) Wodburne, R. T., E. C. Crosby, and R. E. McCotter : The mammalian midbrain and isthmus region. Part II The fiber connections, A The relations of the tegmentum of the midbrain with the basal ganglia in macaca mulatta. *J. comp. Neurol.*, **85**, 67-93, 1946.

和 文 抄 録

痙攣性斜頸に関する実験的研究

京都大学医学部外科学教室第1講座(指導:荒木千里教授)

三 沢 郁 夫

痙攣性斜頸は心因性によつて生ずる場合もあるが、明らかに dystonia musculorum deformans が頸に限局したもの、或は神経系の限局性病変によつて発生したと考えられる場合も少なくない。之等神経系の器質的病変によつて発生した場合、その治療は従来末梢神経に侵襲を加え筋力を弱める方向に向けられて来たが、この方法は侵襲が大きく而も必ずしも効果があるとは限らない。痙攣性斜頸の治療方針があくまで対症的治療であることを考慮すれば現在行なわれつつある定位的大脳基底核破壊により治療せしめうる例も少なくないのではないと思われる。其処で私は猫を用いて(1) 痙攣性斜頸を中枢神経の限局性破壊により作成しうるかどうか？(2) この痙攣性斜頸を大脳基底核破壊で治療せしめうるかどうか？ 若し可能ならばその最も有効な破壊部位は何処か、又どの程度の破壊が必要か？(3) 痙攣性斜頸はどう云う機構で発現するか？等の問題について実験的研究を行ない次の結論を得た。

1) 痙攣性斜頸は中脳被蓋の paramedian を結合腕から赤核小細胞部にわたり positive monopolar electrode で3MA, 10~20V, 20秒間の通電を8ヵ所で行なった電氣的破壊で、102匹中、著明53例(51.0%)、中等度14例(13.7%)、軽度13例(12.7%)、合計80例(77.4%)に発現せしめ得た。著明な痙攣性斜頸を発現した14例を組織学的に検討した処、結合腕交叉部から赤核小細胞部にわたる高さで、一侧の結合腕交叉部、内縦束、網様体内側部の3者が同時に破壊されていたが、一方非発現の7例ではかかる3者同時破壊は1例も認められなかつた。又発現例、非発現例共赤核小細胞部破壊は同程度に認められたので、赤核破壊は痙攣性斜頸発現に特に重要であるとは思われなかつた。

2) 発現した痙攣性斜頸は、睡眠中を除き、頸を中脳被蓋破壊と同側に向け、at rest では tonic な斜頸であるが、何らかの emotional stress が加わると頸を更に

spasmodic に中脳被蓋破壊側に向ける。斜頸の tonic component は破壊後1週間以内に非常によく軽快、或は殆んど消失する。一方 phasic component は1ヵ月以上にわたり殆んど軽快する事はない。中脳被蓋破壊で、痙攣性斜頸の発現例、非発現例共、屢々副症状として、動脈神経麻痺、意識障害、四肢の運動障害を来すことがあるが、之等の症状の強度な例では、痙攣性斜頸は発現し難い様に思われた。働いていない様に思

3) 筋電図上、不規則な筋放電が頸の向きと同側の頸筋群(胸鎖乳突筋、斜角筋、夾板筋、肩胛挙筋、総合筋、二腹筋、下斜筋)により著明に認められ、頸の運動は頸の向きと反対側の頸筋群が電氣的により強くわれる。

4) 上述の中脳被蓋の Paramedian 破壊により生じた痙攣性斜頸を何らの副作用なく軽快或は治療させ得たのは視床破壊した14例中9例で、そのいずれもが Nucleus ventralis の pars posterior medialis a から pars anterior medialis a にかけての部位(大略 V.o.i. に一致)であつた。非軽快の5例ではかかる部位の破壊は認められなかつた。又蒼球を破壊した7例中5例は、何らの副作用なく痙攣性斜頸が軽快或は治療し、そのいずれもが Nucleus entopeduncularis (大略蒼球の内側部に一致)の半分以上が破壊されていたが、非軽快の2例では、この様な程度の破壊は認められなかつた。

5) 健康な猫の中脳被蓋の電氣刺激が第3頸神経、副神経のα運動ノイロンに及ぼす影響は、同側の運動ノイロンの reflex firing を促進させ、一方反対側では抑制させる事を見出した。

6) 痙攣性斜頸発現例で、頸の方向を同側の上部頸神経後根の切断は頸の位置を特に変えなかつたが、反対側での切断は斜頸を消失してしまつた事から、痙攣性斜頸はγ運動ノイロンの不均衡な活動異常もその発現に一役かつていると思われる。